Pediatric Thoracic Actinomycosis

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Abstract

Two cases of pediatric thoracic actinomycosis are presented. A review of the English literature revealed forty-six additional cases in the last 25 years. The mean age of diagnosis was 10 years. The most common presentation was a mass associated with weight loss, fever, and cough. Actinomyces israelii *was the most common causative organism. Prognosis was generally excellent after long-term therapy with penicillin.*

Introduction

Actinomycosis is a rare childhood illness. The three most common sites of actinomycosis are cervicofacial (63%), abdominal (22%), and thoracic (15%), as reported by Cope in 1938.[1] Recent literature has suggested a decline in the incidence of actinomycosis.[2] This has been attributed to increased usage of antibiotics and improved dental hygiene.

The following is a report of two cases of pediatric thoracic actinomycosis that presented to our medical center during the same month. The cases are followed by a review of the English literature for cases of pediatric thoracic actinomycosis reported during the past 25 years.

Case Reports

Case 1

An eight-year-old male presented with one month history of fatigue, weight loss, and a mass on his back, which fluctuated in size and was occasionally painful. Two weeks prior to presentation, he developed fever, productive cough, and intermittent right abdominal and flank tenderness associated with shortness of breath. He also reported feeling short of breath when lying down.

Physical examination revealed a thin and pale male in mild to moderate respiratory distress. Vitals signs: temperature 98.6°F, pulse 161 bpm, respirations 44 /minute, BP 110/66 mm Hg, room air oxygen saturation 99%. Physical exam was remarkable for bilateral non-tender submandibular lymphadenopathy. There were diminished breath sounds over the anterior right upper lung field and tubular breath sounds with dullness to percussion over the posterior right upper lung field. A 10 x 6 cm mass was overlying the right scapula. The mass was slightly tender to palpation, without fluctuance, erythema, or increased warmth.

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Admission laboratory findings included a white blood cell count of 16,600/μL with 87% neutrophils, 4% monocytes, 9% lymphocytes. Hemoglobin and hematocrit were 9.0 g/dL and 28.6%. ESR was 109 mm/hr.

A chest x-ray showed consolidation of the right middle and upper lobes with expansion of the posterior portions of the right 3rd and 4th ribs, along with widening of the left superior mediastinum (Fig 1).

Chest MRI revealed subtle changes involving the upper five thoracic vertebrae with enlargement of the associated paraspinal soft tissue and muscles of the back. There was also extensive consolidation involving the right upper and middle lobes and the medial aspect of the left upper lobe with a small amount of right pleural fluid or thickening (Fig 2).

A biopsy of the mass was obtained, which showed chronic abscess formation and fibrosis with clusters of bacteria compatible with "sulfur granules." Biopsy tissue culture grew *Actinomyces israelii*, *Actinobacillus actinomycetemcomitans*, and *Propionibacterium acnes*.

He was treated with intravenous penicillin for 6 weeks, followed by oral penicillin for 10 months. On follow-up, the patient made a full recovery.

Figure 1

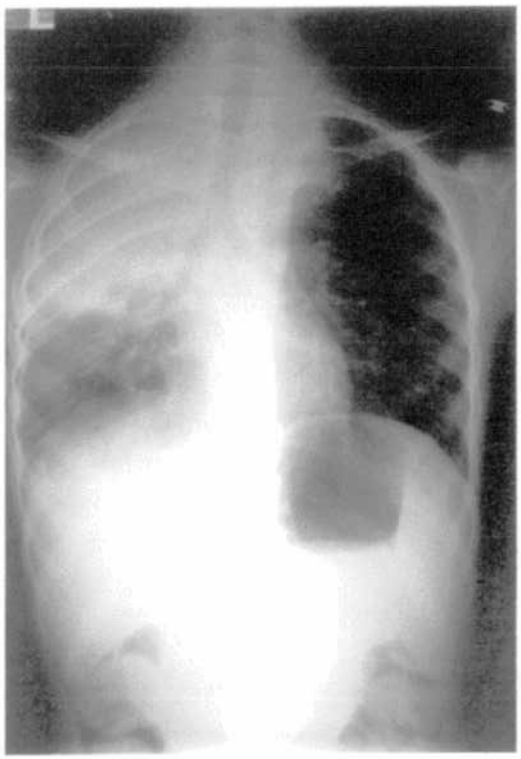
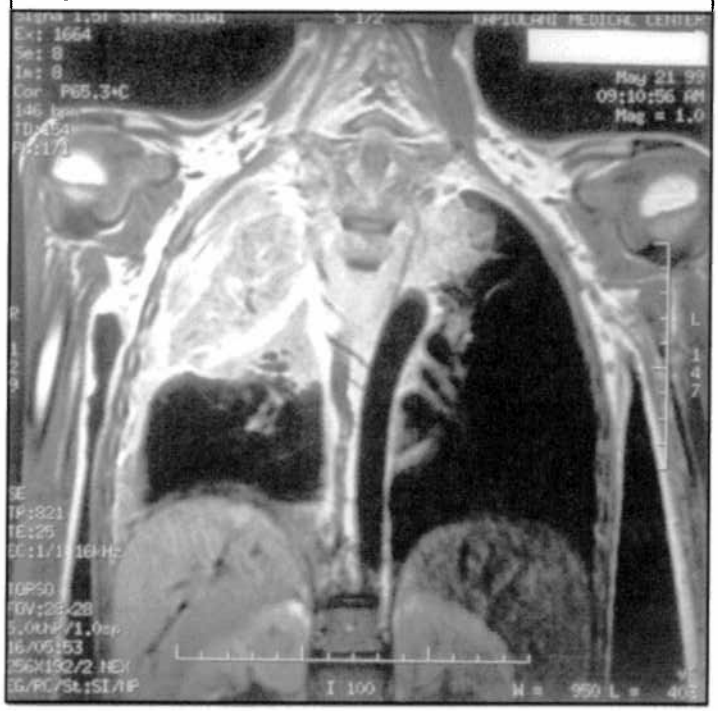


Figure 2



Case 2

A chest x-ray was obtained on an asymptomatic, thirteen year old Filipino female being evaluated for a positive PPD test. The x-ray revealed an extensive left lower lung consolidation with associated sclerosis of the left 7th-11th ribs. Physical examination revealed a thin female in no distress. Pertinent findings included decreased breath sounds over the left lower lung field.

Chest CT showed a heterogeneous enhancing consolidation of most of the left lower lobe with thickening and sclerosis of the overlying ribs. CT guided needle biopsy was not diagnostic. MRI showed a highly vascular left lower lobe mass suggestive of an intralobar sequestration. A left thoracotomy with partial left lower lobectomy was performed and six weeks later the patient developed a moist productive cough.

The left lung mass showed extensive fibrosis with acute and chronic inflammation and focal microabscess formation. A gram stain showed filamentous gram-positive organisms within the bacterial colonies consistent with *Actinomyces*. Culture of the lung mass was negative.

She received 7 weeks of intravenous penicillin, followed by oral penicillin for 12 months. On follow-up, she made a full recovery.

Results

In reviewing the English literature for published cases of pediatric thoracic actinomycosis from 1975 to 2000, thirty-two articles were found reporting a total of 46 cases of pediatric thoracic actinomycosis (2-33).

The patients had a mean age of 10.4 years, with a range of 4.5 to 18 years. Male to female ratio was 2:1, with 30 males and 14 females.

The mean length of illness prior to diagnosis was 5.3 months, with a range of 3 days to 6 years. The most common presenting sign or symptom was a mass (56%), followed by weight loss (48%), cough (37%), and fever (35%). Chest pain was reported in 30%, back pain 15%, and anorexia 20%. A draining sinus or mass was present in 11%. Other common symptoms included shortness of breath, night sweats, hemoptysis, fatigue, malaise, lethargy, and pallor.

Diagnosis was definitively made by culture from a surgical specimen in 33 cases (72%). The most common organism isolated was *Actinomyces israelii* (14 cases). *Actinobacillus actinomycetemcomitans* was reported in 8 cases. A great majority of the cases had multiple organisms. The most common combination was *A. israelii* and *Actinobacillus actinomycetemcomitans* (6 cases). In most cases, a surgical procedure was required for diagnosis. Twenty-one cases were diagnosed after thoracotomy; fourteen were diagnosed by incision and drainage of a mass; eight were diagnosed with needle biopsy. One case was diagnosed by bronchoscopy and one case was diagnosed by culture of a draining sinus.

In this review, 40 of the 46 cases reported were treated with penicillin. Other antibiotics used were erythromycin, clindamycin, ticarcillin, and cephalosporins. Twenty-nine cases specified both type of antibiotic and duration of therapy. The reported durations of antibiotic treatment ranged from 3 weeks to 58 weeks, with a mean duration of therapy of 21.8 weeks. The mean duration of intravenous antibiotic therapy was 4.5 weeks. One patient received oral therapy only, for sixteen weeks.

Outcome was reported in sixteen cases. Fourteen patients showed improvement or complete resolution. One patient was reported to have a complication of systemic-pulmonary arterial fistula and one patient died.

Discussion

Thoracic actinomycosis is usually not considered in the differential diagnosis of pediatric pulmonary disease. Even if considered, confirmation is often difficult because of improper anaerobic culturing, overgrowth of synergistic bacteria, and suppression by prior antibiotic treatment.[11] Some studies suggest an increased incidence in children who are at higher risk for aspiration, who are immune compromised, or who have poor dentition.[12,31] Golden reported that 43% had at least one predisposing factor.[8] However, in this review, only 6 patients had an underlying medical problem (13%). Furthermore, poor dentition was not commonly reported.

The clinical disease of actinomycosis is caused by a species of the genera *Actinomyces*. Once thought to be fungi because of the mycelial appearance of their branching morphology, the etiologic agents of actinomycosis are now clearly identified as true bacteria. *Actinomyces* is a branching filamentous anaeraobic gram-positive rod.

Actinomycosis is most often caused by *Actinomyces israelii* and was identified in 30% of the cases reviewed. Other isolated pathogens included: *A. naeslundii*, *A. viscosus*, and *A. meyeri*. Infection is often polymicrobial with more than one pathogen being identified in 2/3 of the cases. *Actinobacillus actinomycetemcomitans*, the "fellow traveler," is the most common associated pathogen. Other co-pathogens included *Fusobacterium*, *Bacteroides*, *Haemophilus*, *Eikenella*, *Staphylococcus*, *Streptococcus*, *Gamella*, *Neisseria*, and *Eubacterium*.

Actinomycosis is a chronic suppurative infection, which usually results in abscess formation spreading by direct extension without respect for tissue planes. The organisms responsible for actinomy-

cosis are normally found in the oral flora. Thoracic actinomycosis occurs from either aspiration of saliva containing the organisms or more rarely by direct extension from cervicofacial or abdominal actinomycosis. The process often invades the chest wall with formation of a chest wall mass. Sinus tracts may form and extend through overlying skin or to adjacent bones or tissues. These characteristic sinus tracts contain small abscesses and purulent drainage.

Patients may present with a wide range of signs and symptoms. The most typical presentation in cases reported was that of a mass associated with fever, cough, and weight loss. The classic description of a draining sinus was seen in 10%.

Imaging studies are helpful in making a diagnosis. The classic description is a parenchymal infiltrate with extension to the chest wall and erosion of adjacent ribs.[5] The radiologic triad of chronic lower lobe pneumonia, empyema, and wavy periostitis of the ribs, has been described.[10] Extension across interlobar fissures and pleural and pericardial surfaces are described and reflect the ubiquitous nature of the infection. Pleural thickening and pleural effusions are common.

Definitive diagnosis rests on isolating the organism from tissue or pus from a normally sterile body site. True microbiologic identification is commonly not achieved. In this review, 30% had negative cultures. Diagnosis then depends on the clinical picture with identification of the characteristic "sulfur granules" which are tiny lobulated microcolonies of the organisms.

Approximately half the cases (47%) required thoracotomy before the diagnosis was established. Eight cases were diagnosed by needle biopsy. When a soft tissue mass was present, incision and drainage revealed the diagnosis in 10 cases.

Treatment consists of long-term antibiotics. Penicillin is the drug of choice and was used in 87% of the patients. Generally, those receiving other antibiotics had been found to have a penicillin allergy. Length of therapy is not well established. Treatment is generally initiated with parenteral antibiotics, followed by a longer course of oral antibiotics. In this review, the mean length of therapy was 21.8 weeks with 4.5 weeks of parenteral therapy. Oral antibiotics alone may be considered in cases where diagnosis is made early and infection is not widespread.[21]

Thoracic actinomycosis should be considered in the differential diagnosis of pediatric pulmonary disease. The presenting signs and symptoms range from asymptomatic to the classic description of a draining mass with fever, cough and chest pain. Maintaining a high index of suspicion can lead to quicker diagnosis and fewer invasive diagnostic procedures. Generally, prognosis is excellent even in cases of delayed diagnosis.

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